

Preoperative and Postoperative "Aneurysm" Associated With Coarctation of the Aorta

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The reported incidence of aortic aneurysm after surgical repair or balloon angioplasty for aortic coarctation varies widely. To determine the incidence of aneurysm formation after surgery, preoperative and postoperative cineangiograms from 65 patients who underwent operation at age 1.5 ± 3.4 years were examined. Repair included a prosthetic patch in 14 patients, end to end anastomosis in 28 and subclavian flap in 23. Aneurysm was documented by change in contour or irregularities in contour at the repair site or by abnormal dimensions at the repair site, defined by the ratio of the widest measurement at the repair site to the measurement of the aorta at the diaphragm.

An aneurysmal bulge above the ductus diverticulum was identified in 14 (23%) of 60 patients preoperatively; the area

showed no change 4.72 ± 4.07 years after surgery. Significant changes at the repair site were seen in only three patients, all of whom had Dacron patch repair. One patient had a change in contour at the repair site, one had an abnormally high repair site to diaphragmatic aorta ratio and one had a progressive increase in this ratio. Thus, during childhood years, 3 (5%) of 65 patients were diagnosed as having aneurysm at the surgical repair site.

In conclusion, 1) comparison with preoperative cineangiograms, especially for aneurysmal bulges above the ductus arteriosus, is essential before an aneurysm can be attributed to coarctation repair by any technique, and 2) aneurysm developed only in patients subjected to Dacron patch repair.

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Since the first report (1) of repair of coarctation of the aorta, surgery has been the primary therapy for this form of congenital heart disease. Aneurysm formation has been reported as a complication of surgical repair; long-term follow-up studies (2-8) show a prevalence rate of 5% to 27%. Every patient reported to have aneurysm formation after surgical repair underwent repair with prosthetic patch material. Aneurysm formation has also been reported (9-13) in up to 55% of patients undergoing balloon angioplasty. Thus, aortic integrity before and after surgical repair of coarctation must be evaluated in relation to aneurysm formation.

To better define the phenomenon of aneurysm formation, cineangiograms from 65 consecutive patients with coarctation who required preoperative and postoperative studies for various cardiac lesions were reviewed for 1) presence of aneurysmal changes at the coarctation site before surgical repair; 2) appearance of new geometric changes suggestive of aneurysm formation at the repair site after surgery; and 3) comparison of measurements of the aorta at the repair site and diaphragm.

Methods

Patient characteristics. Preoperative cineangiograms were available in 60 of 65 patients; 108 postoperative studies of the 65 patients were reviewed. Preoperative cineangiograms could not be located in two patients and were not considered valid in three because of technical problems. Surgical repair was performed over an age range of 0.1 to 15 years (mean 1.56 ± 3.41). Postoperative studies, obtained 5.7 ± 4.8 years (range 2 months to 9.5 years) after the coarctation repair, were performed to evaluate associated cardiac lesions in 46 patients, suspected recoarctation in 7 patients and the results of intracardiac repair for congenital heart disease in the other patients. Serial postoperative studies, performed 4.5 ± 2.4 years (range 2 months to 9.5 years) apart, were available in 31 patients; 7 patients had more than two postoperative studies. In patients who had more than one postoperative study, the initial and the most recent postoperative study were used for serial comparison.

Analysis of cineangiograms. Cineangiograms obtained in the left anterior oblique projection were analyzed by two of the investigators (S.R.P. and R.A.H. or S.R.P. and D.A.G.) without prior knowledge of the surgical repair. Each cineangiogram was reviewed for visual delineation of any bulge or irregularity in aortic contour at or around the repair site. Measurement of the widest dimension of the repair site was compared with measurement of the aorta at the diaphragm (Fig. 1); this ratio was termed the repair site ratio. Measurements were also obtained from preoperative cineangiograms

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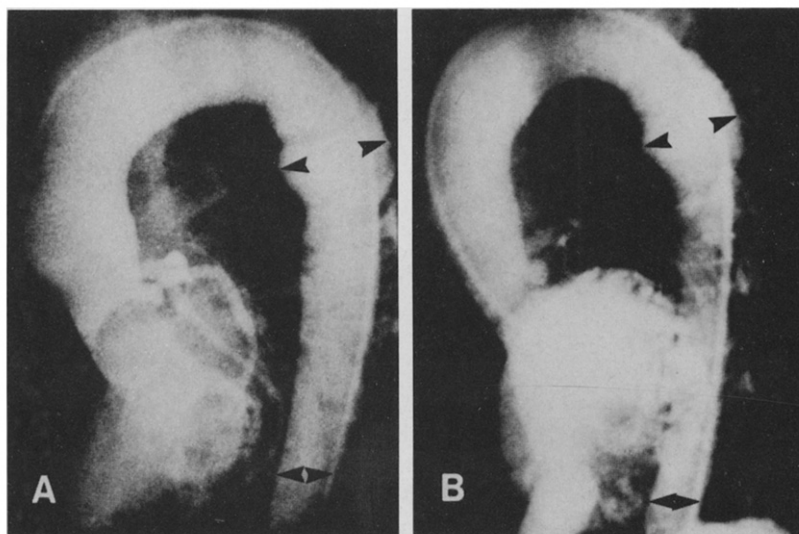


Figure 1. The repair site ratio. The widest dimension at the coarctation repair site (**upper arrowheads**) was compared with measurement of aorta at the diaphragm (**lower arrowheads**) in the left anterior oblique projection. This patient had an exceedingly high ratio (1.8) 1 year after the repair (**A**). The ratio was unchanged 2.5 years after repair (**B**).

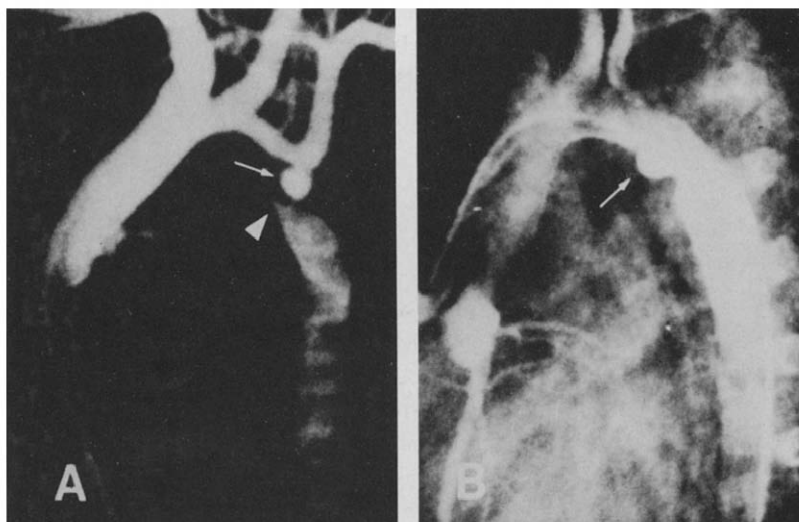


Figure 2. Preductal bulge on preoperative (**A**) and postoperative (**B**) cineangiograms. The preductal bulge is shown by the **arrows** and the ductus arteriosus by the **thick arrowhead**. The patient underwent subclavian flap repair; no change was seen in the contour of the preductal bulge postoperatively.

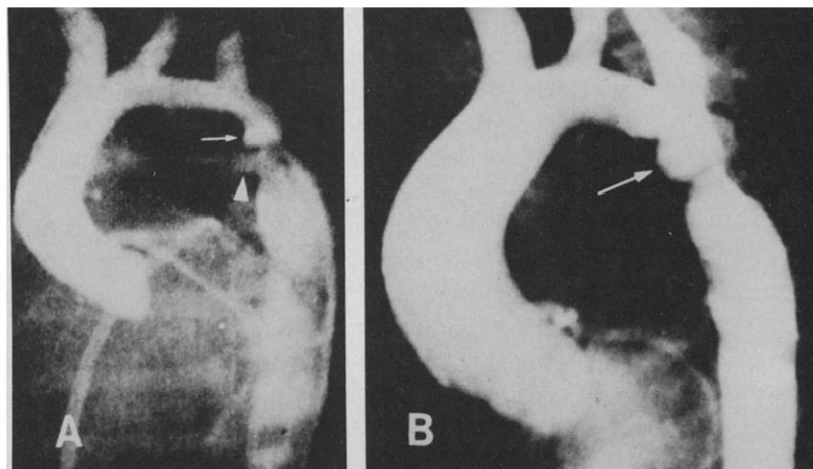
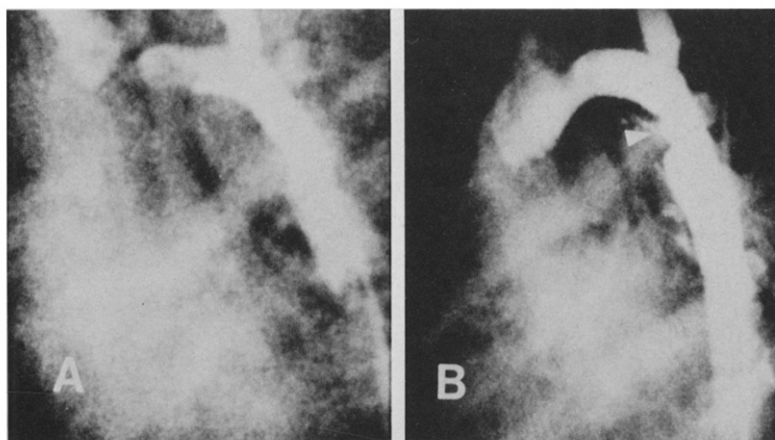


Figure 3. Preductal bulge on preoperative (**A**) and postoperative (**B**) cineangiograms. The preductal bulge (**arrow**) and the ductus arteriosus (**arrowhead**) are seen as distinctly separate structures on the preoperative angiogram.

Figure 4. Occurrence of a new geometric change at the site of coarctation repair. **A**, Cineangiogram recorded 4 years after coarctation repair. **B**, A new spherical bulge (arrowhead) appeared on the angiogram 8 years after repair.



at the coarctation site, diaphragmatic aorta and just proximal to the left subclavian artery. All measurements were made directly from an opaque glass screen attached to a Vanguard cine film projector. Because ratios were examined, attenuation in size resulting from cineangiographic projection was not a factor. Measurements for the widest diameter at the repair site and diaphragmatic aorta were obtained from different frames in some of the patients, depending on the quality of opacification. In patients who had more than one postoperative cineangiogram, comparison of the repair site ratio among various types of surgical repair was completed using measurements from the initial postoperative cineangiogram.

Statistical analysis. Data were analyzed with use of the Kruskal-Wallis test and Spearman rank correlation test; a p value ≤ 0.05 was considered significant. Data are expressed as mean values \pm SD.

Results

Presence of aneurysmlike areas preoperatively: occurrence of "preductal bulges." Discrete areas of bulging in the anterior aortic wall were noted above the ductus arteriosus in 14 (23%) of the 60 preoperative cineangiograms (Fig. 2 and 3). These areas were best seen in the left anterior oblique projection and were not well seen in the right anterior oblique projection. The aneurysmal area disappeared after end to end repair of the coarctation but persisted after Dacron patch repair or subclavian flap repair. Patients who had this finding preoperatively underwent postoperative cineangiographic studies 4.7 ± 4.07 years (range 6 months to 9 years) after surgery that showed no significant change in the size or contour of the affected area.

Incidence of Postoperative Aneurysms

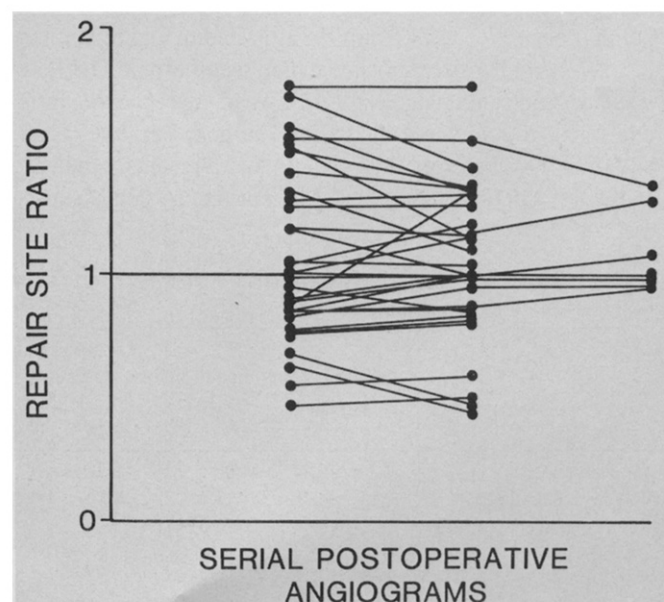
Aneurysm formation was identified in 3 (5%) of the 65 patients with use of the following criteria.

Occurrence of new geometric changes at the repair site (one patient). A discrete spherical bulge was seen in the left anterior oblique projection in one patient and was believed

to represent aneurysm formation (Fig. 4). The bulge appeared postoperatively and was not seen on preoperative angiograms. This patient underwent Dacron patch repair. This change was not accompanied by a significant change in aortic measurements at the repair site.

Repair site ratio > 1.68 (one patient). Repair site dimension was compared with diaphragmatic aortic dimension on each of the postoperative cineangiograms (Fig. 1). The 14 patients who underwent Dacron patch repair were studied 5.84 ± 4.21 years (range 6 months to 12 years) after surgery and had a postoperative repair site ratio of 1.08 ± 0.31 (range 0.5 to 1.8). The repair site ratio in 28 patients who underwent end to end repair was 1.06 ± 0.25 (0.6 to 1.7). These patients underwent cineangiography 7.7 ± 5.2 years (range 9 months to 17.9 years) after surgery. The 23 patients who had subclavian flap repair were studied 2.5 ± 1.9 years (range 2 months to 6.6 years) after repair and had a ratio of 1.19 ± 0.30 (range 0.7 to 1.8). No statistical difference was seen in

Figure 5. Serial postoperative repair site ratios in 32 patients. A significant progressive increase in the ratio is noted in one patient.



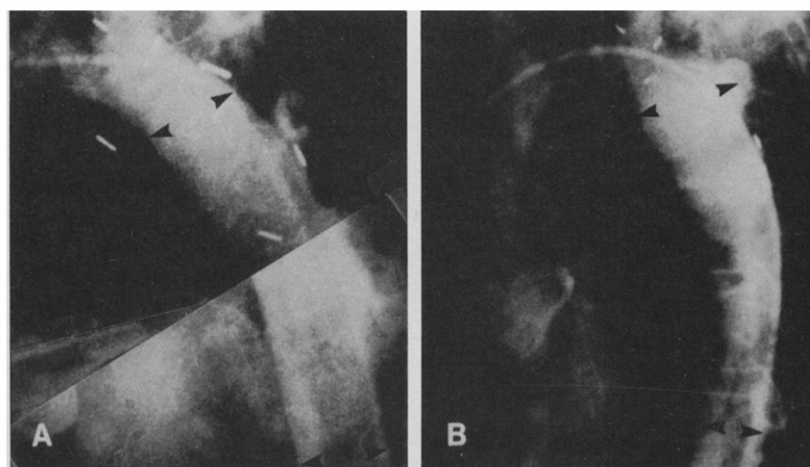


Figure 6. Increase in repair site ratio on serial postoperative follow-up. **A**, Composite from an angiogram obtained 6 months after surgical repair. **B**, Angiogram 10 years later. The ratio increased from 0.95 to 1.43. Opacification from a bony prominence is superimposed on the lateral aspect of the aorta at the repair site, making the aneurysmal bulge appear larger than marked.

the repair site ratio among the three techniques of coarctation repair ($p = 0.28$, Kruskal-Wallis test). The mean repair site ratio from all 108 postoperative cineangiograms was 1.06 ± 0.31 (range 0.5 to 1.8). From these measurements one patient with a ratio of 1.8 was diagnosed as having aneurysm formation. This patient had undergone Dacron patch repair.

Increasing repair site ratio in serial postoperative studies (one patient). Serial postoperative studies were completed 4.5 ± 2.4 years (range 2 months to 9.5 years) after the initial postoperative study in 31 patients. Six of these patients had Dacron patch repair, 16 had end to end anastomosis and 9 had subclavian flap repair. The final ratio of 1.1 ± 0.31 was not significantly different from the initial ratio of 1.05 ± 0.36 ($p = 0.3$) (Fig. 5). The ratio remained constant or changed marginally ($\pm 10\%$) in 19 patients, declined by 10% to 30% in 8, increased by 15% in 2 and increased by 50% in 1 patient (Fig. 5 and 6). This increase of 50% in 1 patient was considered to represent aneurysm formation. The patient had undergone Dacron patch repair.

Residual gradients. The residual gradient during postoperative catheterization was 23.4 ± 22.2 mm Hg after Dacron patch repair, 17.3 ± 16.5 mm Hg after end to end repair and 8.5 ± 7.2 mm Hg after subclavian flap repair (Table 1). There was no significant linear relation between the repair site ratio and residual gradients ($r = 0.23$). The postoperative repair site ratio was therefore not predictive of a residual gradient. In the three patients identified as having aneurysmal changes

at the coarctation repair site, the gradients were 0, 6 and 20 mm Hg, respectively.

Discussion

Preductal bulges. Aneurysm formation at the repair site has been documented (2-13) after surgical coarctation repair using prosthetic patches and after balloon angioplasty for coarctation. Experimental studies (14) suggest that extensive surgical resection of intima with or without patch aortoplasty predisposes to aneurysm formation. A similar mechanism of intimal interruption is believed responsible for aneurysm formation after balloon angioplasty (15). We showed that aneurysmal bulges may be present in the aorta above the ductus before any form of repair. Discrete bulges or bumps were present above the ductus arteriosus preoperatively in 23% of our patients and could be identified as separate from the ductus arteriosus. Although discrete aneurysmlike bulges below the coarctation site have been reported (13), to the best of our knowledge an aneurysmal bulge above the ductus has not been previously identified. The preductal bulge may be secondary to increased blood pressure above the coarctation site coupled with intrinsic weakness in the aortic wall due to the fragile periductal tissue. Follow-up of our patients over a mean of almost 5 years after surgery showed no change in the size and contour of these areas. The preductal bulges seemed to disappear after resection and end to end anastomosis but persisted after Dacron patch repair or subclavian flap angioplasty (Fig. 7).

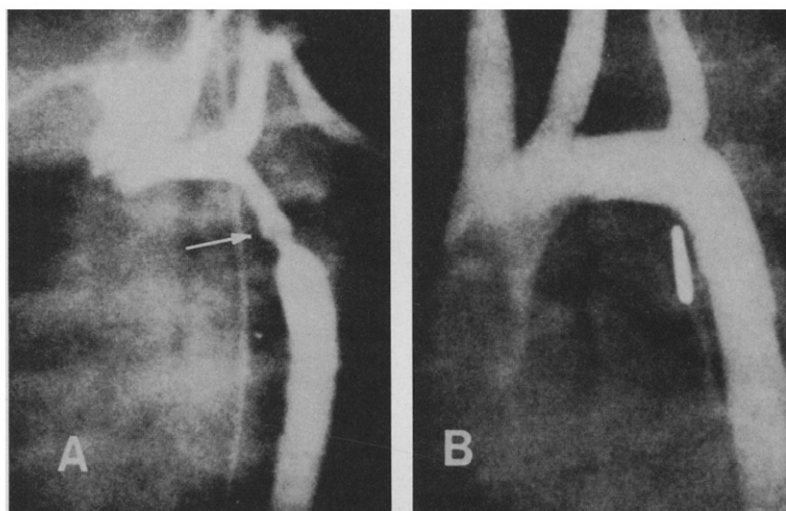
Therefore, before specific conclusions can be reached about development of new aneurysms after any form of coarctation repair, preoperative cineangiograms must be reviewed. The presence of ductal diverticula has been described (16). We observed irregularity in the aortic wall secondary to the presence of a ductus diverticulum in two of our patients (Fig. 8). However, the presence of a preductal bulge is a finding different from that of ductal diverticulum.

New geometric changes at the repair site. One of the criteria we used to define aneurysm was the appearance of

Table 1. Repair Site Ratio Versus Residual Gradient at Catheterization in 65 Patients

| | Dacron Patch Repair (n = 14) | End to End Repair (n = 28) | Subclavian Flap Angioplasty (n = 23) |
|---------------------------|------------------------------------|----------------------------------|---|
| Repair site ratio | 1.08 ± 0.31 | 1.06 ± 0.25 | 1.09 ± 0.30 |
| Follow-up period (yr) | 5.8 ± 4.2 | 7.7 ± 5.2 | 2.5 ± 1.9 |
| Residual gradient (mm Hg) | | | |
| Mean | 23.4 ± 22.2 | 17.3 ± 16.5 | 8.5 ± 7.2 |
| Range | 0-70 | 0-60 | 0-22 |

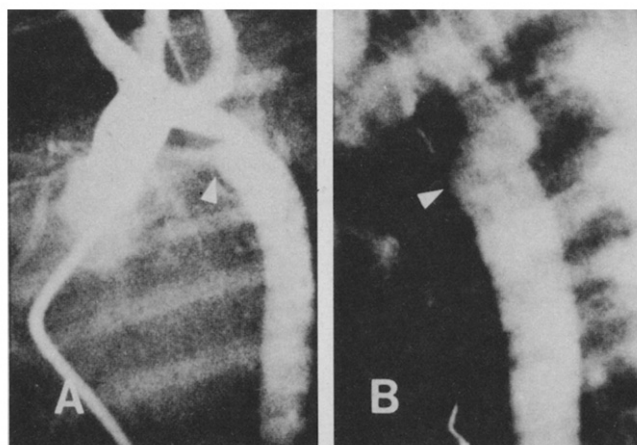
Figure 7. A, Preoperative angiogram showing pre-ductal bulge (arrow). B, Disappearance of the bulge after an end to end anastomosis for coarctation repair.



new geometric changes at the site of coarctation repair. This occurrence was noted as an appearance of a discrete new bulge at the repair site in one patient (Fig. 4). Absence of significant widening in such patients makes it likely that the aneurysmal change would be missed on a chest radiograph.

Repair site ratio. The ratio of widest diameter at the repair site to diameter at the diaphragmatic aorta was chosen in an attempt to reach an objective measurement for aneurysm identification. Had we chosen an absolute size to reflect aneurysmal change, variations in magnification among various patients could have been a problem in data interpretation. A similar attempt was made in an earlier study (8) and a ratio ≥ 1.5 was considered to suggest aneurysmal dilation after patch angioplasty repair. Patients who had coarctation repair by other surgical techniques were not included in that study. Other, earlier studies (2-7) that reported aneurysm formation after patch angioplasty did not use aortic measurements for the diagnosis of an aneurysm.

Figure 8. Ductal diverticulum. A, Preoperative cineangiogram shows a moderately large ductus arteriosus and a bulge in the aortic wall (arrowhead) in the area of the ductus. B, The ductal bulge persisted postoperatively (arrowhead).



The ratio tends to be larger, although not statistically significantly so, after subclavian flap repair; this probably reflects a redundancy, because the ratio often has a progressive decrease with growth. Serial measurement of the ratio in 31 patients showed a decrease in 8 patients and a significant increase in only 1 patient. It is therefore difficult to judge one particular observation on the repair site ratio as being consistent with an aneurysm because the temporal relation to the surgical repair is important and must be considered. Such was the case in the earlier attempt (8) made to define an aneurysm as an aortic ratio ≥ 1.5 . Of the 29 patients in that series who underwent patch angioplasty, 7 were identified as having an aneurysm. However, no serial follow-up was available on these seven patients. When we pooled our data from all 105 postoperative observations of the repair site ratio, the mean ratio was 1.06 ± 0.31 . The mean value ± 2 SD would statistically comprise at least 95% of the ratio values seen at various periods of time after all types of repair. A repair ratio ≥ 1.68 at any time would therefore be highly suggestive of an aneurysmal change regardless of the mode of repair.

Because geometry of the aorta at repair site is not normal, we intentionally avoided comparing the repair site ratio with normal control values. Caution should be exercised when attempting to dichotomize the repair site ratio so that all values beyond a certain point could be considered abnormal. The wide range of repair site ratios would make any form of dichotomy arbitrary. On the basis of progressive increase in ratio on serial postoperative follow-up or a repair site ratio ≥ 1.68 , aneurysm was diagnosed in two patients in our series. Both of these patients had Dacron patch angioplasty.

The calculation of a ratio in itself involves uncertainty; edge detection on cineangiograms could be difficult, and a slight overestimation of the size of the repair site and underestimation of the size of the descending aorta will yield a spuriously high value, leading to a mistaken diagnosis of aneurysm. We therefore believe that serial evaluation is more important in reaching the diagnosis of an aneurysm. An

increase in the ratio seen on serial evaluation by any imaging modality (cineangiography, echocardiography or nuclear magnetic resonance imaging) is probably more important than an isolated, single value of an increased ratio.

Implications. Thus, during a mean of 5.7 ± 4.8 years after surgical coarctation repair, the overall prevalence rate of aneurysm was 5% in our series (3 of 65 patients). All three patients had Dacron patch repair. Previous estimates of aneurysm formation range from 5% to 24%. Indeed, if the incidence of aneurysm is as high as 24% after surgical coarctation repair and the incidence is shown to be lower after balloon angioplasty on long-term follow-up, angioplasty may become the treatment of choice for coarctation repair. However, aneurysms have not been frequently reported on long-term follow-up after resection and end to end anastomosis or subclavian flap repair. Subsequent surgery for aneurysms after primary correction of coarctation by surgery or balloon angioplasty could be associated with increased risks due to the decrease in collateral circulation, thereby increasing the possibility of spinal cord ischemia (17).

Residual gradients. The repair site ratio, which reflects aortic remodeling after surgery, had no correlation with the residual gradient measured at the time of postoperative cardiac catheterization. This is partly because although the ratio was lower in some patients because of a narrow repair site, a significant gradient was not found because good collateral circulation was present. A smaller value for repair site ratio was therefore not always indicative of a large residual gradient. Although the gradient was generally smaller after subclavian flap repair than that for other types of repair, the period of follow-up was shorter. A residual gradient after subclavian flap repair could progressively worsen, a possibility that could not be examined in our study. The development of aneurysmal changes in three patients was not related to a high residual gradient; their gradients ranged from 0 to 20 mm Hg.

Conclusions. On the basis of cineangiographic analysis before and after coarctation repair, we conclude that the overall incidence of aneurysm after long-term follow-up of surgical coarctation repair is 5%. Aneurysms developed only when Dacron patch repair was used. No new aneurysmal change was found at the coarctation repair site in 51 patients after end to end repair or a subclavian flap procedure. Several factors must be considered when contemplating a diagnosis of aneurysm associated with coarctation repair: 1) a discrete bulge above the ductus arteriosus may be present preoperatively and should not be mistaken for postoperative

aneurysm; 2) appearance of a new bulge at the repair site may suggest the development of an aortic aneurysm; and 3) a repair site to diaphragmatic aorta ratio ≥ 1.68 or a progressive increase in the ratio suggests aneurysmal change.

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